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The prevalence of ankyloglossia in 302 newborns with breastfeeding problems and sucking difficulties in Barcelona: a descriptive study

ABSTRACT

Aim *Oral and maxillofacial development is influenced by the lingual frenulum and also affects breathing, occlusion, sucking, swallowing, speech, among others. Ankyloglossia in the newborn may result in breastfeeding difficulties: maternal nipple pain and/or erosion or mastitis, poor weight gain and excessively long breastfeeds. The main objective of this work is to study the prevalence of ankyloglossia in newborns with breastfeeding difficulties.*

Material and Methods *This is a transversal descriptive study of 302 patients, between 0 and 6 months, who attended the hospital as a result of breastfeeding difficulties. All patients with sucking problems and ankyloglossia were included in this study and followed the multidisciplinary treatment protocol made up of the services of Breastfeeding, Speech Therapy and Orofacial Rehabilitation and Oral and Maxillofacial Surgery.*

Results *1,102 newborns were seen at the paediatric service of Hospital de Nens, Barcelona (Spain) during 2 years; 302 had breastfeeding difficulties and of these, 171 were diagnosed with ankyloglossia (60 girls and 111 boys). Coryllos Grade 3 ankyloglossia was the most prevalent (59.6%) type; 85 infants (49.7%) were exclusively breastfed and 26 (50.35%) were mixed fed (formula and breastfeeding). Only 43 patients had a family history of tongue-tie (25.1%).*

Conclusions *Ankyloglossia linked to breastfeeding difficulties must be treated by a multidisciplinary team. We have found a high prevalence of the condition since the population studied are newborns with sucking problems. If a frenotomy is necessary, we recommend stimulating suction with myofunctional therapy before and after surgery to avoid scar retraction.*

Keywords *Ankyloglossia; Breastfeeding; Frenotomy; Myofunctional therapy; Sucking problems; Tongue-tie.*

Introduction

The lingual frenulum is a membrane that connects the tongue to the floor of the mouth and influences the growth and development of the oral and maxillofacial cavity affecting breathing, occlusion, suction, swallowing and speech [Queiroz Marchesan, 2004; Martinelli et al., 2012].

The ability to suck is innate in all mammals; humans learn the swallowing-suction pattern while still in uterus. This learning begins at week 16 of gestation with the swallowing reflex, and is complemented at week 20 with the sucking reflex. The lingual frenulum is an embryonic residual tissue that originates in the stages of development of the oral cavity and, on occasions, there is an incorrect division of the genioglossus and hyoglossus muscles, leaving the frenulum outside the tongue [Amir et al., 2006; Marchesan, 2012].

Ankyloglossia is defined as a limitation of protrusion and elevation of the tip of the tongue due to the shortness of the frenulum and/or genioglossus muscles [Olivi et al., 2012; Jamilian et al., 2014; Crippa et al., 2016]. It

FIG. 1 Coryllos anatomical classification.

can be observed in different age groups with different treatment indications for each group [Segal et al., 2007]; ankyloglossia in newborns and ankyloglossia during infancy and adolescence [Coryllos et al., 2004; Hogan et al., 2005; Dollberg et al., 2006; Olivi et al., 2012; Ferrés-Amat et al., 2016a].

Mothers and infants may face a variety of difficulties in mastering breastfeeding: a breastfeeding mother may experience sore nipples, blocked ducts, nipple and breast infection, and signs of low milk supply. Infants may be unable to transfer milk adequately, in part due to their inability to maintain an effective latch and seal onto the breast [Griffiths, 2004; Kotlow, 2011; Haham et al., 2014].

Ineffective sucking can cause breast pain to the mother, poor weight gain the newborn and excessively long feeds [Coryllos et al., 2004; Ferrés-Amat et al., 2016b].

When the baby is born, the mandible is in the posterior position relative to the upper maxilla, giving the newborn a convex profile (physiological retrognathia). During breastfeeding, stimulation of the buccal musculature occurs and the structures of the stomatognathic apparatus of the newborn are activated. Breastfeeding is a decisive factor in the newborn's growth, and there is scientific evidence of the benefits of this form of feeding as opposed to the bottle since the required muscular movements for extraction of milk during breastfeeding is crucial for the optimal growth of the lower third of the face [Marchesan, 2004; Sanchez-Molins et al., 2010; Martinelli et al., 2012].

At the Hospital de Nens, Barcelona (Spain) we have created a Suction Pathology Unit (CELERE), formed by the Breastfeeding Service, Speech Therapy and Orofacial Rehabilitation Service, and Oral and Maxillofacial Surgery Service for the study, treatment, and follow-up of newborns with sucking problems. The main objective of this study is to describe the prevalence of ankyloglossia in newborns with breastfeeding problems.

Material and methods

We conducted a descriptive cross-sectional study of infants with sucking disorders during lactation and diagnosed with ankyloglossia.

The study population consists of all patients between 0 and 6 months of age who, for a period of two years, are referred by the paediatric service to the Suction Pathology Unit (CELERE) and are diagnosed with ankyloglossia associated with inefficient suction.

The diagnosis of ankyloglossia is carried out according to the Coryllos classification which defines the following four types of frenulum.

- Type I: Fine and elastic frenulum; the tongue is anchored from the tip to the alveolar ridge and it is found to be heart-shaped.
- Type II: Fine and elastic frenulum; the tongue is anchored 2-4 mm from the tip to almost near the alveolar ridge.
- Type III: Thick, fibrous, non-elastic frenulum; the tongue is anchored from the middle of the tongue to the floor of the mouth.
- Type IV: The frenulum cannot be seen, but palpated; it has a fibrous and/or thick and shiny submucous anchoring from the base of the tongue to the floor of the mouth [Coryllos et al., 2004; Martinelli et al., 2012].

Besides the Coryllos classification (Fig. 1), functional-clinical criteria were also taken into account: poor weight gain (less than 100 grams per week), excessively long breastfeeds (more than 60 minutes) and maternal pain (breastfeeding should not be a cause of pain for the mother).

In order to evaluate the incidence and the severity of the alteration of suction presented by newborns, a classification based on the severity of the condition was used, combining three parameters to be studied with the degree of ankyloglossia:

- Mild: Coryllos Type III-IV and clinical criteria (excluding pain).
- Moderate: Coryllos Type I-II and clinical criteria or either Coryllos Type III-IV and 2 clinical criteria or with just pain.
- Severe: Coryllos Type I-II plus 2 or 3 clinical criteria, or either Coryllos Type III-IV and 3 clinical criteria.

The Visual Analogue Scale (VAS) was used in the evaluation of pain. Weight gain was assessed by recording the child's weigth before each breastfeeding and stimulation session, using the same digital scale. The duration of the breastfeeds was recorded by the mother herself, and to this end, the following duration times were established: <15 minutes, 15-30 minutes, 30-60 minutes, >60 minutes; the value chosen for the study and evaluate

FIG. 2 Surgical Technique: Lingual Frenotomy.

the evolution was the highest time.

The variables studied were: the age of the infant at the first visit, once the lactation problem was detected in the baby-mother binomial, sex of the baby, mother's age, type of pregnancy and delivery, type of feeding (exclusive or mixed breastfeeding), duration of breastfeeds, weight gain of the patient, family history of ankyloglossia, and the number of infants diagnosed with ankyloglossia and treated by the multidisciplinary CELERE team.

Criteria for inclusion: patients younger than 6 months, healthy ASA I, without a diagnosis of systemic disease or syndrome. Criteria for exclusion: patients whose parents did not sign the informed consent to be part of the study.

A complete physical examination of the baby was performed with special attention to muscle tone and the anatomy of the oral cavity and maxillofacial region. The newborn was examined for: lingual frenulum, type of delivery, gestation age, the early onset of breastfeeding, the quantity of milk sucked by the newborn, the use of soothers and/or bottles.

The mother was assessed for the following: maternal pain during the feeds using the "Visual Analogue Scale" (VAS), adjustments in changes of position and grip, transfer of milk and a physical examination of the breasts.

The treatment undertaken following the procedure established by the Suction Pathology Unit (CELERE) is as follows: if ineffective sucking continues after 3 sessions of breastfeeding, the lactation specialist refers the infant to the service of myofunctional therapy (MFT) where the rooting and sucking reflexes are stimulated through extraoral and intraoral exercises by performing four 30-minute sessions, during one month. If a lingual frenotomy is decided, the treatment is undertaken using a grooved catheter and a 12cm Metzenbaum dissecting scissors (Fig. 2). After surgery, stimulation and breastfeeding are performed.

A statistical analysis of the data was carried out. Categorical variables are summarised with the frequency and the percentage, and the numerals with the median [minimum; maximum] or with the mean (standard deviation). Analyses were performed with software R: A language and environment for statistical computing. R Foundation for Statistical Computing, Vienna, Austria. Version 3.2.1.

Results

During the study period at the Service of Paediatrics, 1,102 first visits were made to infants less than 6 months old. A total of 302 patients (128 girls and 174 boys) attended the Breastfeeding Service due to suction difficulties.

171 patients with ankyloglossia (60 girls and 111 boys) less than 6 months old were enrolled in the study and classified according to the treatment received: all the mothers received recommendations for postural correction during lactation. If the problems persisted, patients were then treated with myofunctional therapy, and if the symptoms were not corrected then, they underwent surgery.

Patients were divided into the following groups:

1. Breastfeeding Sessions Group (BFS): In 33 patients (18 girls and 15 boys) (19.3%, IC 95%= 13.7%; 26.0%), suction problems were solved with the Breastfeeding Sessions (SLM), just by correcting the position while breastfeeding.
2. Group that undertook Myofunctional Therapy (MFT) and Breastfeeding Sessions: MFT was carried out in 50 cases (16 girls and 34 boys) (29.2 %, IC95%= 22.5 %; 36.7 %).
3. Group that had surgery (MFS), in addition to BFS and MFT. A total of 88 infants (26 girls and 62 boys) underwent a frenotomy (51.5 %, IC 95%= 43.7 %; 59.2 %). There were no surgical complications.

In the three groups and in the total sample, improvements in the effectiveness and comfort of breastfeeding were observed. Figure 3 shows the treatment applied according to the severity of ankyloglossia.

The characteristics of the three groups are described in Table 1. The prevalence of ankyloglossia in our study group was 15%. We highlight a prevalence of 64.9% of ankyloglossia in males, and 37.1% had a caesarean delivery. As regards breastfeeding, we observed prior to the intervention of the CELERE team that 50.3% of the mothers practised mixed breastfeeding, which decreased to 12.3% after treatment. Type 3 ankyloglossia was the most frequently lingual frenulum type, found in 59.6% of the infants, followed by type 2 in 26.9% and type 4 in 13.5% of cases. The duration of the feeds between

30-60 minutes before the treatment by the CELERE team was 37.4%, followed by 36.3% of over 60 minutes. After the intervention of the CELERE team, feeds of less than 30 minutes were the most prevalent in 91.8% of the cases and only 8.24% of patients took 30-60 minutes (Figure 4). Figure 5 shows the weight gain before and after treatment and Figure 6 describes pre-treatment and post-treatment pain.

Only 14% of the mothers had previous experience in breastfeeding. Forty-three infants (25.1%) were found to have a first-degree family history of ankyloglossia: parents or siblings.

	All	only BFS	BFS+MFT	Surgery	p-valor
	N=171	N=33	N=50	N=88	
Infant's gender: Boy	111 (64.9%)	15 (45.5%)	34 (68.0%)	62 (70.5%)	0.032
Mother's age	35.0 (3.47)	34.5 (3.72)	35.7 (3.11)	34.8 (3.55)	0.240
Days from birth to initial consultation	29 [0;268]	25 [4;73]	30 [5;268]	30 [0;177]	0.421
Type of pregnancy:					0.696
Spontaneous	148 (86.5%)	28 (84.8%)	42 (84.0%)	78 (88.6%)	
IVF/assisted	23 (13.5%)	5 (15.2%)	8 (16.0%)	10 (11.4%)	
Type of delivery:					0.263
Vaginal	107 (62.9%)	24 (72.7%)	33 (66.0%)	50 (57.5%)	
Cesarean	63 (37.1%)	9 (27.3%)	17 (34.0%)	37 (42.5%)	
Gestational age	39.3 (1.41)	39.6 (1.32)	39.3 (1.29)	39.2 (1.50)	0.341
Breastfeeding pre-treatment					0.032
Maternal	85 (49.7%)	17 (51.5%)	32 (64.0%)	36 (40.9%)	
Mixed	86 (50.3%)	16 (48.5%)	18 (36.0%)	52 (59.1%)	
Breastfeeding post-treatment					0.191
Maternal	150 (87.7%)	31 (93.9%)	46 (92.0%)	73 (83.0%)	
Mixed	21 (12.3%)	2 (6.06%)	4 (8.00%)	15 (17.0%)	
Coryllos:					<0.001
Type 1-2	46 (26.9%)	5 (15.2%)	8 (16.0%)	33 (37.5%)	
Type 3	102 (59.6%)	19 (57.6%)	31 (62.0%)	52 (59.1%)	
Type 4	23 (13.5%)	9 (27.3%)	11 (22.0%)	3 (3.41%)	
Pre-treatment pain	5.03 (3.08)	4.12 (2.67)	5.10 (3.27)	5.33 (3.07)	0.153
Post-treatment pain	0.84 (1.30)	0.70 (1.16)	0.98 (1.46)	0.81 (1.25)	0.597
Pre-treatment breastfeeding time:					0.029
<30 min	45 (26.3%)	11 (33.3%)	14 (28.0%)	20 (22.7%)	
30-60 min	64 (37.4%)	15 (45.5%)	23 (46.0%)	26 (29.5%)	
>60 min	62 (36.3%)	7 (21.2%)	13 (26.0%)	42 (47.7%)	
Post-treatment breasfeeding time:					0.142
<30 min	156 (91.8%)	30 (90.9%)	49 (98.0%)	77 (88.5%)	
30-60 min	14 (8.24%)	3 (9.09%)	1 (2.00%)	10 (11.5%)	
>60 min	0 (0.00%)	0 (0.00%)	0 (0.00%)	0 (0.00%)	
Pre-treatment weight gain					0.571
<100 gr/sem	109 (63.7%)	23 (69.7%)	27 (54.0%)	59 (67.0%)	
100-200 gr/sem	37 (21.6%)	6 (18.2%)	14 (28.0%)	17 (19.3%)	
>200 gr/sem	25 (14.6%)	4 (12.1%)	9 (18.0%)	12 (13.6%)	
Post-treatment weight gain:					1.000
<100 gr/week	3 (1.75%)	0 (0.00%)	1 (2.00%)	2 (2.27%)	
100-200 gr/week	165 (96.5%)	33 (100%)	48 (96.0%)	84 (95.5%)	
>200 gr/week	3 (1.75%)	0 (0.00%)	1 (2.00%)	2 (2.27%)	
Post-treatment weight gain					1.000
Gain 100+	168 (98.2%)	33 (100%)	49 (98.0%)	86 (97.7%)	
Gain <100 gr/sem	3 (1.75%)	0 (0.00%)	1 (2.00%)	2 (2.27%)	
Previous breastfeeding	24 (14.0%)	2 (6.06%)	10 (20.0%)	12 (13.6%)	0.205
Family history of ankyloglossia	43 (25.1%)	7 (21.2%)	12 (24.0%)	24 (27.3%)	0.772

TABLE 1 Participants general characteristics and related to the therapeutic group.

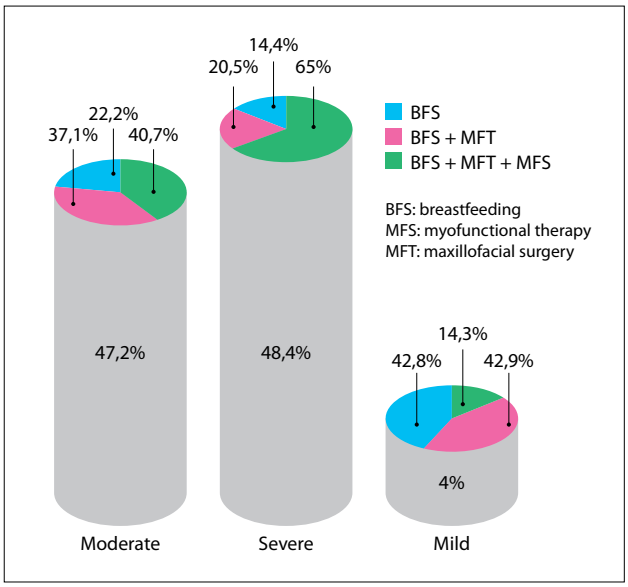

FIG. 3 Treatment applied according to the severity of ankyloglossia.

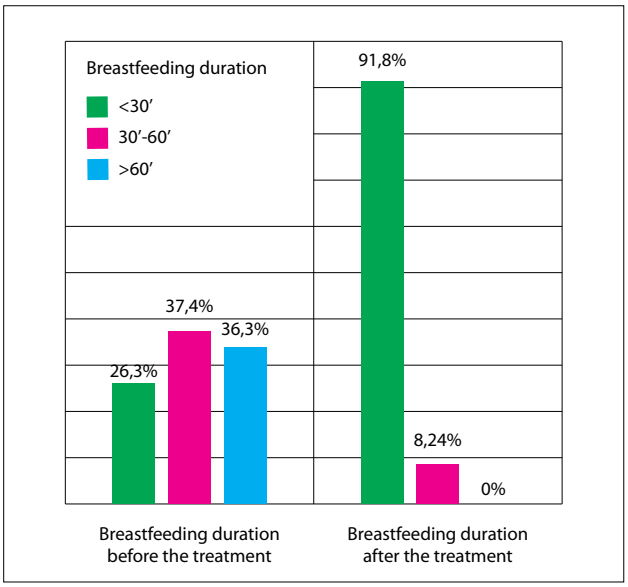

FIG. 4 Duration of the feeds before and after the treatment.

Discussion

The prevalence of ankyloglossia in infants is variable in the literature, reflecting the lack of a consistent definition. Estimates range from 4.2% to 10.7% in newborns (Rowan-Legg, 2015). The prevalence of ankyloglossia in our study group was 15.5%. This is rather high compared to what has been published in other articles since the total population we have studied are newborns whose families attend a specialised service in Human Lactation, mostly due to problems during breastfeeding. In 2014 González Jiménez et al. published a 12% prevalence of ankyloglossia in the general population of Asturias, a value that is 2 to 3 times higher than that published by other authors such as Messner et al., who in the year 2000 found a prevalence of 4.8%, and Ricke, with a prevalence of 4.2% in 2005 [Messner et al., 2000; Ricke et al., 2005; González Jiménez et al., 2014].

In our sample, we found 25.1% of newborns with a first-degree family history of ankyloglossia and a prevalence of 64.9% of males, similar to the percentage (62% males) found in the study by González Jiménez et al. [2014]. The hypertrophic lingual frenulum is mostly found in males due to their genetic characteristics [Messner et al., 2000; Hogan et al., 2005; Ricke et al., 2005].

In some cases, the need to quantify the degree of ankyloglossia with a mathematical formula in which different measures have to be taken makes the quorum of the diagnosis difficult for professionals [Messner and Lalakea, 2000; Grandi, 2012; Marchesan, 2012; Martinelli et al., 2012; Cuestas et al., 2014]. In other cases, frenulums needing surgery were considered, omitting all others. Others lead to confusion by defining the severity

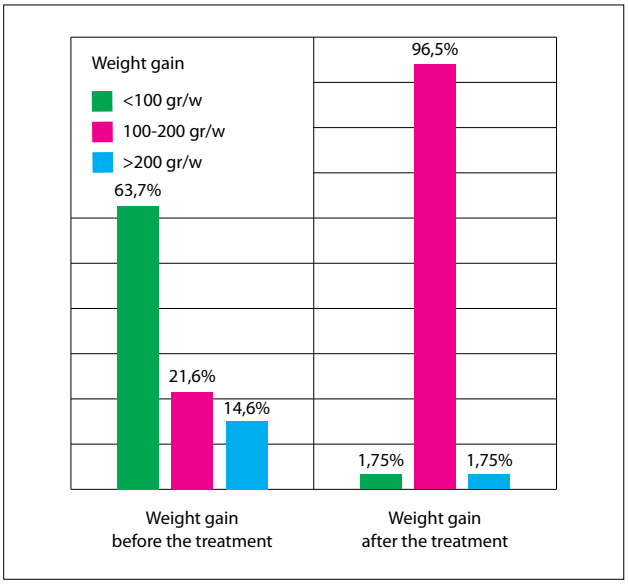

FIG. 5 Weight gain before and after treatment.

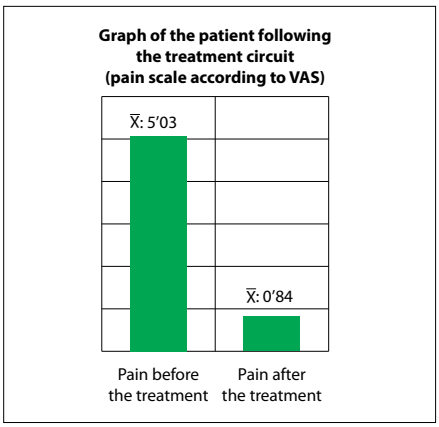

FIG. 6 Pain before and after treatment.

of the frenulums conversely to the numbering given to them [Amir et al., 2006; Edmunds et al., 2012; Webb et al., 2013; Haham et al., 2014; Junqueira et al., 2014].

One of the main unsolved problems of ankyloglossia is its classification and diagnosis. There is a great deal of controversy due to the lack of a universally accepted definition and objective diagnostic criteria and, on the other hand, to the variability of the symptomatology presented by both mothers and babies, since there is no relation between the severity of ankyloglossia and breastfeeding difficulties. As we have observed, these difficulties may be partially offset by maternal or infant conditions that make it difficult to identify ankyloglossia as a cause of breastfeeding problems [Rowan-Legg, 2004; Kupietzky and Botzer, 2005; Dollberg et al., 2006; Martinelli et al., 2015].

The classification of Coryllos permits to identify type III and IV frenulums, which can go unnoticed to the naked eye, by means of palpation [Coryllos et al., 2004; González Jiménez et al., 2014]. For the diagnosis of ankyloglossia in newborns it is necessary to combine functional and anatomical criteria [González Jiménez et al., 2014; Ferrés-Amat et al., 2016b]. Haham et al. [2014] did not find any statistical correlation between the Coryllos type of lingual frenulum and the presence of lactation problems.

The Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF) has been developed to provide a quantitative evaluation of the lingual frenulum and it is recommended when performing a frenotomy [Amir et al., 2006; Edmunds et al., 2012; Olivi et al., 2012]. There are validation studies that combine anatomical and functional criteria [Amir et al., 2006; Ingram et al., 2015].

In our study, we used the Coryllos classification as a means of diagnostic criteria associated with the limitation of lingual mobility with difficulties in suction and latching onto the breast, assessing: pain-cracks-mastitis in the mother, poor weight gain of the newborn and excessively long feeds (more than 60 minutes). In the study by González Jiménez et al. [2014], according to the Coryllos classification, type II was the most frequent (54%). In our study, type III is the most frequent lingual frenulum, found in 59.6% of cases.

Lingual frenotomy is a treatment that health professionals frequently recommend for treating ankyloglossia and breastfeeding problems [Martinelli et al., 2015]. When the association between ankyloglossia and the main problems linked to breastfeeding are clearly identified, surgical intervention is considered necessary and the frenotomy should be performed by an experienced specialist [Rowan-Legg, 2015].

From our results, we observed that in the total of 33 patients, suction problems were managed and solved just with the Breastfeeding Sessions, through correcting the position when breastfeeding. In 50 cases, the sucking problems were solved with Myofunctional Therapy (without surgery) applying extraoral and intraoral stimulation to recover the rooting and sucking reflexes. It is vital to detect the sucking problems in the early days following birth to begin immediate stimulation and correct immature suction. A frenotomy was performed on 88 patients, and no surgical complications were recorded.

Based on the available evidence, frenotomy cannot be recommended for all infants with ankyloglossia, since there is no absolute relationship between ankyloglossia and breastfeeding difficulties [Rowan-Legg, 2015]. Most babies with ankyloglossia are asymptomatic and have no feeding problems. In cases of ankyloglossia and significant breastfeeding difficulties, there is some evidence that frenotomy can improve feeding (Ricke et al., 2005; Francis et al., 2015; Rowan-Legg, 2015; Ferrés-Amat et al., 2016b).

A thorough intraoral examination, including inspection of the tongue and its function, should be performed in newborns, particularly in case of feeding difficulties [Rowan-Legg, 2015]. We would like to point out the importance of the initial assessment of the problem by an expert in breastfeeding, in our case, an International Board Certified Lactation Consultant (IBCLC) paediatrician, to detect associated difficulties, and performing an initial correction, knowing that most breastfeeding problems are due to poor posture and grasping, and the timely referral to myofunctional therapy and surgery when required. MFT should be carried out by an expert therapist to avoid relapses and the persistence of poor mobility of the oral and the perioral musculature [Ferrés-Amat et al., 2016b]. Surgery (frenotomy) should be undertaken for the complete release of the tongue [Francis et al., 2015].

In treating ankyloglossia, the choice of treatment should only be made after careful assessment. The paediatrician should coordinate the collaboration among the various specialists involved (paediatric dentist, oral surgeon and speech and orofacial myofunctional therapist) [Olivi et al., 2012; Crippa et al., 2016; Ferrés-Amat et al., 2016b].

Scientific evidence suggests that frenotomy can be associated with some improvements in breastfeeding and in the pain suffered by mothers when breastfeeding, but these are small, short-term studies with incompatible methodology, and are rather limited and insufficient [Rowan-Legg, 2015]. More studies are necessary but for the moment, based on current available evidence, frenotomy cannot be recommended for all babies with ankyloglossia [Edmunds et al., 2013; Emond et al., 2014; Francis et al., 2015].

The results of our study show a higher incidence of severe cases treated by the three services that make up our unit; a result that demonstrates that the incidence in suction alterations is directly related to the most important disorders, such as poor weight gain, excessively long breastfeeds and maternal pain. The fact that there is no need for treatment or insufficient clinical evidence justifies the lower incidence breastfeeding problems in the group classified as mild. Therefore, it is unnecessary for the whole team to intervene and the alteration can

be resolved simply through correcting posture and/or stimulation.

Conclusions

Suction alterations can condition the child's orofacial growth; through restoring suction, we avoid possible malocclusions that could occur during craniofacial development and may compromise other oral functions.

The prevalence of ankyloglossia in our study group is high given that the population we studied were newborns with breastfeeding problems visited at a specialised lactation unit. Early detection of the causes of suction problems and the timely treatment are crucial to achieve optimal breastfeeding.

Alterations in the lingual frenulum can be corrected and surgery is not necessary in all cases, as some can be resolved with myofunctional therapy, improving their grip thus avoiding more traumatic interventions and achieving better results in feed times and comfort for the mother when breastfeeding. If a frenotomy is necessary, myofunctional therapy is recommended before and after to stimulate suction, in order to minimise scar tissue retraction.

Finally, it is worth emphasising the importance of teamwork and the need to unify diagnostic criteria to solve problems in the field of breastfeeding.

Conflict of interests

The authors declare that they do not have any conflict of interest. This study has been performed by the Research Group of the Fundació Hospital de Nens de Barcelona.

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